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Development and face validity of a cerebral visual impairment motor questionnaire for children with cerebral palsy

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Abstract

Aim The objectives of this study were (i) to develop two cerebral visual impairment motor questionnaires (CVI-MQ's) for children with cerebral palsy (CP): one for children with Gross Motor Function Classification System (GMFCS) levels I, II and III and one for children with GMFCS levels IV and V; (ii) to describe their face validity and usability; and (iii) to determine their sensitivity and specificity.

Backgrounds The initial versions of the two CVI-MQ's were developed based on literature. Subsequently, the Delphi method was used in two groups of experts, one familiar with CVI and one not familiar with CVI, in order to gain consensus about face validity and usability. The sensitivity and specificity of the CVI-MQ's were subsequently assessed in 82 children with CP with (n = 39) and without CVI (n = 43). With the receiver operating curve the cut-off scores were determined to detect possible presence or absence of CVI in children with CP.

Results Both questionnaires showed very good face validity (percentage agreement above 96%) and good usability (percentage agreement 95%) for practical use. The CVI-MQ version for GMFCS levels I, II and III had a sensitivity of 1.00 and specificity of 0.96, with a cut-off score of 12 points or higher, and the version for GMFCS levels IV and V had a sensitivity of 0.97 and a specificity of 0.98, with a cut-off score of eight points or higher.

Conclusion The CVI-MQ is able to identify at-risk children with CP for the probability of having CVI.

Keywords
cerebral palsy, cerebral visual impairment, occupational therapists, paediatric physical therapist, screening

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Introduction

Cerebral visual impairment (CVI) is the major cause of visual impairment in developed countries (Good *et al.* 1994; Ortibus *et al.* 2011; Liew *et al.* 2015). Approximately 30% of children diagnosed with various forms of cerebral palsy (CP) also suffer from CVI (Schenk-Rootlieb *et al.* 1993; Dutton and Jacobson, 2001; Stiers *et al.* 2002; Da Costa *et al.* 2004; Ghasia *et al.* 2008).

The spectrum of visual impairments in children with CP is broad and includes forms of ocular visual impairment (OVI) such as strabismus, reduced visual acuity, ocular nystagmus and CVI, which is a problem of central origin. (Da Costa *et al.* 2004; Dutton and Jacobson, 2001; Ghasia *et al.* 2008; Schenk-Rootlieb *et al.* 1993, Stiers *et al.* 2002). CVI ranges from no light perception to normal visual acuity and, in the presence of cognitive visual dysfunction, a visual processing disorder that leads to misinterpretation of the visual world (Edmond and Foroozan, 2006).

Cerebral visual impairment is a result from impaired processing of visual information in the presence of a (nearly) intact ophthalmological system (corrected vision >0.3 and/or field of vision >30°) (Goodale and Milner, 1992; Dutton and Jacobson, 2001; Dutton *et al.* 2004; Fazzi *et al.* 2004, 2012). With regard to the controversy and heterogeneity of the diagnosis of classification of CVI, in our study, CVI is defined in terms of visual deficits of any likely cerebral cause, thereby including a wide range of visual sensory and visual perceptive deficits of known neurological underpinnings, and excluding visual deficits due to optical abnormalities. Children with CP and CVI develop more slowly in the areas of self-care, mobility and social function than children with CP without CVI (Da Costa *et al.* 2004; Ghasia *et al.* 2008). Detecting CVI in children with CP using motor questionnaire as an additional tool can help the paediatric physical therapists and occupational therapists to make a better estimation of motor ability in a child, and to adapt therapy via manual and verbal support.

Paediatric physical therapists and occupational therapists are often the first professionals to assess and treat children with CP. This puts them in a position to identify red flags for CVI (at risk of having a visual impairment). Such red flags allow professionals to review the impact of CVI on the observed motor behaviour and to ensure the identification of signs and symptoms of CVI in children with CP. Red flags instruments for CVI are lacking; it is important to develop a CVI screening tool to identify these signs.

Children with CP have many limitations, which hamper a thorough standardized assessment of motor functioning, and the current assessments do not account for the presence of visual impairments in children with CP. Hence, professionals have to rely on observations or findings from the child's history to diagnose CVI (Ortibus *et al.* 2011; Salavati *et al.* 2014).

A motor screening tool consisting of items related to the contribution of visual perception to perform a motor activity is therefore needed. Thus far, no validated CVI screening tool is available to screen children with CP and to identify the possible contribution of CVI on motor impairment (Dutton and Jacobson, 2001; Ortibus *et al.* 2011; Dutton *et al.* 2012).

Gross motor function of children with CP can be classified into five different severity levels using the Gross Motor Function Classification System (GMFCS) (Rosenbaum *et al.* 2007). Because of these large functional differences at the level of motor disability, we decided to develop two different CVI Motor Questionnaires (CVI-MQ): the CVI-MQ for children with CP with GMFCS I, II and III and CVI-MQ for children with GMFCS IV and V. The content of the CVI-MQ for children with GMFCS levels I, II and III includes motor items for children about higher motor skills such as walking, stair-climbing and jumping, while the CVI-MQ for children with GMFCS levels IV and V contains motor skills such as rolling over and reaching.

Our aims were first to develop two CVI-MQs for children with CP, second to describe their face validity and usability, and third to determine their sensitivity and specificity to detect a possible presence of CVI in children with CP.

Methods

The study was conducted in three phases. First, based on existing literature and according to the GMFCS levels, we developed two CVI-MQs for children with CP: one for GMFCS levels I, II and III and one for GMFCS levels IV and V. Second, the Delphi method was used to gain consensus about the face validity and to gain insight into the usability of the two MQs among a panel of experts working with CP-CVI children and a panel of experts working with CP children (Gracht von der, 2012). The purpose of the study and required procedures was explained to both groups, and they were subsequently asked for consent to participate in the study. Following consent, the experts were asked about their age, profession and working experience. The predetermined goal for the usability was to reach a consensus of 95% agreement in experts that were not familiar with CVI. Third, data of children with CP with and without CVI were used to calculate sensitivity and specificity of the two CVI-MQs to detect the probability of presence or absence of CVI in children with CP and to determine cut-off scores.

The two CVI-MQs measure the degree of presence or absence of CVI in children with CP, where higher scores indicate higher levels of probability to predict CVI in children with CP. The sum score of each CVI-MQ represents the sum items perceived by the therapist being present at a certain child. Each CVI-MQ counts the number of CVI items. If an item was considered not applicable by the therapist, then this item was not added to the sum score.

Phase 1 – developing CVI-MQs

We searched for literature published between May 1995 and December 2015 using the PubMed, PsychLit, EMBASE, PEDro and MEDLINE databases using Medical Subject Headings (MeSH) terms and text words. The following queries were used: 'Cerebral Palsy' [MeSH] *AND*/*OR* 'Cerebral Visual Impairment' *OR* 'Cortical Visual Impairment' *OR* 'Cortical blindness' *OR* 'vision disorder' [MeSH], in combination with *AND* 'Gross Motor Classification System I, II, III, IV, V' *AND* 'motor activity' *OR* 'functional skills' *OR* 'self-care' *OR* 'mobility' *AND* 'screening' *OR* 'observation' *OR* 'questionnaire'.

The multiple-choice items in the two CVI-MQs were selected on the basis of the current questionnaires used by (i) the home intervention team for children with CVI at Royal Dutch Visio and Bartiméus (centres of expertise for blind and visually impaired people in the Netherlands); (ii) the visual skills inventory available from the studies of Dutton *et al.* (Dutton and Jacobson, 2001; Dutton *et al.* 2004, 2012; Dutton, 2013); (iii) literature reviews of features of CVI in children; (iv) the adapted version of the paediatric evaluation of disability inventory, Dutch version (PEDI-NL); and (v) the Gross Motor Function Measure-88 (GMFM-88) for children with CVI (Haley *et al.* 1992; Russell and Rosenbaum, 2002; Fazzi *et al.* 2004; Edmond and Foroozan, 2006; Dutton, 2013; Salavati *et al.* 2015a,b).

Phase 2 – adaptation of CVI-MQs

Adaptation

The Delphi method was used among a panel of 19 CP and CVI (CP-CVI) experts and a panel of 20 CP experts. The Delphi method was applied in a sequence of sequential questionnaires or 'rounds', interspersed by controlled feedback, in order to seek the most reliable consensus of opinion from a purposeful sample of experts (Powell, 2003; Gracht von der, 2012). In this study, face validity was defined as an opinion of CP-CVI experts on the CVI-MQs. We therefore asked the CP-CVI experts whether or not the CVI-MQs measured presence or absence of CVI in children with CP.

To investigate the face validity of developed CVI-MQs, first we invited a group of CP-CVI experts of Royal Dutch Visio and Bartiméus by e-mail. To assess usability of CVI-MQs, we also invited a group of CP experts by posting an invitation on the website of their organization and by e-mail. In this study, the usability was defined as usage of the CVI-MQs by professionals not familiar with CVI. The CP experts worked at private practices and healthcare practices.

Data collection for adaptation

Firstly, the CP-CVI experts gave their comment on the content of questionnaires. Feedback information could be written about any items, and the experts were specifically asked whether each item was appropriate for children with CVI. If not, we asked what needed to be added or changed to make it appropriate.

The predetermined goal was to reach an experts' consensus of 65% on each item after the first round, 75% after the second round and 85% after the third round for agreement with each item as well as content of the CVI-MQs (Powell, 2003).

First Delphi round

The CP-CVI experts gave comments individually on the content of each item. We asked these experts, which items needed to be changed or added to the two CVI-MQs and why.

Second and third Delphi rounds

After receiving the comments of CP-CVI experts, we processed all of the suggestions in the questionnaires and resubmitted them twice to these experts. We asked them whether the content of each item and instruction part of CVI-MQs was appropriate for children with CVI and how long it took them to answer the items on each CVI-MQ.

After the third Delphi round, we asked the CP experts to comment on the two CVI-MQs. We asked CP experts whether the items and the instruction part of CVI-MQs were clearly stated and how long it took them to answer all the items on each CVI-MQ.

Phase 3 – sensitivity and specificity of CVI-MQs

Children with any type of CP with and without CVI were recruited from Royal Dutch Visio and allied healthcare practices. Inclusion criteria were presence of CP and CVI, mild or moderate intellectual disability, and age at testing of

the CVI-MQ for children between 4 and 16 years. Level of intellectual disability was derived from the children's medical files. Children with hearing difficulties (>30 dB hearing loss) and severe or profound intellectual disability (IQ < 40) were excluded. Diagnosis and classification of CP were extracted from the children's medical files and verified by a rehabilitation specialist. Based on possible effects on motor functioning and to detect potential confounding characteristics, we also collected data on gender and prevalence of epilepsy and speech/language development

The diagnosis of CVI was based on the children's medical files, which were determined by ophthalmological and psychological/neuropsychological assessments and by assessment data reported by a developmental coach specialized in working with children with visual impairments. Except for the diagnosis of CVI, there was no additional information available about the criteria of CVI for using in this study.

The study was approved by the Medical Ethical Committee (METc-2015-048) of University Medical Center Groningen (UMCG), Groningen, the Netherlands. Written informed consent was obtained from the children's parents.

Statistical analyses

Data were analysed using Statistical Package for Social Sciences (SPSS, IBM Corp., Armonk, NY, USA), v.22 software. We used the Receiver operating curve (ROC) depicting of sensitivity versus 1-specificity (1 – true positive proportion) for different values of the cut-off point. The area under the curve (AUC) represented an overall accuracy measured covering all possible interpretation thresholds. An area of 0.9–1.0 represented an excellent value for a test, a value between 0.8 and 0.9 is good, between 0.7 and 0.8 fair, between 0.6 and 0.7 poor and between 0.5 and 0.6 fail. AUC values closer to 1 are preferable (Eng, 2005). An optimal cut-off point was determined with sensitivity and specificity rates set at good value (0.8–0.9). We analysed the CVI-MQs data to investigate their predictive value to predict the presence of CVI in children with CP. Sensitivity and specificity of the two CVI-MQs were analysed from children with CP, with and without CVI. Because the CVI-MQs were meant to identify at-risk children with CP for the probability of having CVI, and to refer for full diagnosis, it was important to obtain high sensitivity so as to miss the fewest possible number of cases. The diagnostic accuracy was evaluated by the positive predictive value, negative predictive value, positive likelihood ratio, negative likelihood ratio and confidence interval of both CVI-MQs. We also created scatter plots to visualize the distribution of CVI-MQ measurements for children with CP, separately for CVI is present and absent. To report percentage of agreement, we gave this percentage on each item of the two CVI-MQs.

Results

Adaptation of CVI-MQs

Nineteen health experts familiar with CP and CVI (CP-CVI experts) participated in the development of the two CVI-MQs; five occupational therapists, 13 paediatric physical therapists and one a behavioural scientist. Mean (SD) age of the experts was 51 (10) years and their mean years (SD) of experience with children with CP and CVI was 20 (9). All experts worked at Royal Dutch Visio and Bartiméus.

In addition, to determine usability of two CVI-MQs, 20 health experts familiar with CP but not familiar with CVI (CP experts) participated in the development of the two questionnaires; sixteen of them were paediatric physical therapists and four occupational therapists. Their mean (SD) age was 46(11) years, and mean years (SD) of experience with children with CP was 19 (11). All of them worked at health care centres and private practice.

First Delphi round

On the CVI-MQ for children with GMFCS I, II and III (Table 1), 68% of the CP-CVI experts agreed about the content of items 5, 6, 9, 10, 12, 17, 18, 20, 21 and 24–27 (agreement percent 74–89%). Most comments were about items 1–4, 7, 8, 11, 13–16, 19, 22 and 23 (agreement percent 42–63%). For example, they suggested that for item 3 it is important to add the word 'moved' to the item: 'The child bumps into moved toys or furniture when it belly-crawls'. Item 5 was changed by deleting 'the child has difficulty without verbal support' from the item, 'The child has difficulty anticipating differences in height when it walks, for example when stepping down from the sidewalk onto the road'. With respect to stairs (items 7–8, Table 1), the experts suggested having two separate items, one for climbing stairs and another for walking down stairs.

On the CVI-MQ for children with GMFCS IV and V (Table 2), 71% of the CP-CVI experts agreed about items 5–8, 9–12 (agreement percent 71–89%). The experts suggested combining two items by deleting 'talking at the same time' from item 8 'The child bumps into obstacles/persons when driving a wheelchair (mechanic/electric)', to make it suitable for children with CVI. Most experts' comments were about items 1–4, 13–14 (agreement percent 53–63%). They suggested that by adding information on instruction those items would be suitable for children with CVI. The experts suggested adding the item 'The child grabs an object if it produces sound' (item 13, Table 2).

Table 1. Cerebral Visual Impairment Motor Questionnaire (CVI-MQ) for children with Cerebral Palsy (CP), GMFCS I, II and III

Item	CVI-MQ for children with GMFCS I, II and III	First Delphi round (%)	Second Delphi round (%)	Score		
				YES (%)	NO (%)	Not applicable (%)
Gross motor skills						
1	The child belly-crawls if stimulated by movement*, sound production*, fluorescence*, high-contrast* toys, or verbal support*.	53	89.5	70	23	7
2	The child crawls if it is stimulated by movement*, sound production*, fluorescence*, high-contrast* toys, or verbal support*.	47	89.5	74	23	3
3	The child bumps into moved toys or furniture when it crawls.	63	100	44	54	2
4	The child is more uncertain when it walks in an unfamiliar environment compared with a familiar environment.	53	100	51	49	0
5	The child has difficulty anticipating differences in height when it walks, for example when stepping down from the sidewalk onto the road.	74	94.7	56	42	2
6	The child walks slower in unfamiliar environments.	79	94.7	56	44	0
7	The child will walk up an unfamiliar staircase one step at the time, always leading with the same foot, whereas it will walk up a familiar staircase with alternating feet at each step.	42	94.7	54	37	9
8	The child will walk down an unfamiliar staircase one step at the time, always leading with the same foot, whereas it will walk up a familiar staircase with alternating feet at each step.	47	94.7	54	37	9
9	The child bumps into obstacles/persons when it walks.	74	94.7	44	56	0
10	The child bumps into obstacles/persons when it runs.	74	94.7	44	47	9
11	The child walks significantly slower when there is no person to follow.	63	94.7	44	54	2
12	The child hesitates when it moves from one room to another; this occurs when the child both leaves and enters a room.	79	100	44	54	2
13	The child falls* and/or trips* over obstacles.	58	100	49	44	7
14	The child does not jump off an elevated platform.	53	89.5	46	33	21
15	The child does not jump forwards*, sidewards* or backwards*.	53	89.5	44	35	21
16	When catching a ball, the child misses a non-sound-producing* and/or non-fluorescent*, non-high-contrast* ball more often than a sound-producing*, fluorescent*, high-contrast* ball.	63	100	44	51	5
17	The child kicks behind/next to the ball when kicking a non-sound-producing*, non-fluorescent*, lower-colour*/-contrast* ball.	89	100	44	49	7
18	The child rolls*/throws* a ball towards a person if there is verbal support.	79	89.5	56	39	5
19	The child has difficulty estimating the distance and speed of other road users.	58	100	58	35	7
20	The child has difficulty finding the route to the class or the school playground when walking at school.	68	100	44	54	2
Fine motor skills / Reaching and grasping						
21	The child reaches behind/bumps into small objects. The child only grabs the object after touching it.	84	100	44	54	2
22	The child manipulates the toy with its hands instead of exploring it with its eyes.	63	100	51	49	0
23	The child has difficulty copying figures with a pencil.	58	100	30	42	28
24	The child reaches more precisely when reaching for moving objects.	74	100	47	51	2
25	The child reaches more precisely towards sound-producing*, high-contrast*, fluorescent*, illuminating* objects compared with non-sound-producing*, non-high-contrast*, non-fluorescent*, non-illuminating* objects.	89	100	47	51	2

Continues

Table 1. (Continued)

Item	CVI-MQ for children with GMFCS I, II and III	First Delphi round (%)	Second Delphi round (%)	Score		
				YES (%)	NO (%)	Not applicable (%)
26	The child does not reach for and look at an object at the same time.	89	100	47	53	0
27	The child reaches towards toys but has difficulty finding the toys in a crowded background. For example, finding a block on a full table or in a basket filled with toys.	84	100	47	51	2

Results of percentages consensus experts after first and second Delphi round. Percentage of agreement for test result of sensitivity and specificity on each item: 'Yes', 'No', 'Not applicable'
**Circle as applicable*

Table 2. Cerebral Visual Impairment Motor Questionnaire (CVI-MQ) for children with Cerebral Palsy (CP), GMFCS IV and V

Item	CVI-MQ for children with GMFCS IV and V	First round Delphi (%)	Second round Delphi (%)	Score		
				YES (%)	NO (%)	Not applicable (%)
Gross motor skills						
1	The child turns it head to follow, if encouraged by sound production*, fluorescence*, high-contrast* toys or verbal stimulation.	58	94.7	64	31	5
2	The child lifts its head when lying on its stomach, if encouraged by sound production*, fluorescence*, high-contrast* toys or verbal stimulation.	63	94.7	64	31	5
3	From a sitting position the child lifts its head, if encouraged by sound production*, fluorescence*, high-contrast* toys or verbal stimulation.	63	94.7	64	31	5
4	The child belly-crawls if encouraged by sound production*, fluorescence*, high-contrast* toys or verbal stimulation.	58	94.7	42	28	30
5	The child bumps into moved toys or furniture when it belly-crawls.	74	100	31	36	33
6	The child crawls/belly-crawls slower in an unknown environment with the same surface as a known environment.	89	94.7	28	36	36
7	The child has difficulty finding the route to the class or school playground when driving a wheelchair (mechanic/electric).	79	100	36	51	13
8	The child bumps into obstacles/persons when driving a wheelchair (mechanic/electric).	71	100	43	44	13
Fine motor skills/ reaching and grasping						
9	The child reaches more precisely for moving objects than for non-moving objects.	84	94.7	51	49	0
10	The child reaches more precisely for sound-producing*, high-contrast*, fluorescent*, illuminating* objects than for non-sound-producing*, non-high-contrast*, non-fluorescent*, non-illuminating* objects.	79	100	51	49	0
11	The child looks away when it grabs an object.	79	100	54	46	0
12	The child reaches for a toy but has difficulty finding the toy in a crowded background. For example, finding a block on a full table or in a basket filled with toys.	84	100	54	46	0
13	The child grabs an object if it produces sound.	53	89.5	64	36	0
14	The child explores*/manipulates* toys with its mouth or hands instead of exploring it with its eyes.	53	89.5	46	54	0

Results of percentages consensus experts after first and second Delphi round. Percentage of agreement for test result of sensitivity and specificity on each item: 'Yes', 'No', 'Not applicable'
**Circle as applicable*

Second Delphi round

Ninety-seven percent of the CP-CVI experts agreed on the content of items of the CVI-MQ for children with GMFCS I, II and III (Table 1), and 96% agreed on the items of the CVI-MQ for children with GMFCS IV and V (Table 2). Because of the high percentage of agreement, we sent both CVI-MQs to CP-CVI experts as well as to the CP experts.

On the CVI-MQ for children with GMFCS I, II and III (Table 1), the highest level of agreement (100%) among CP-CVI experts was for items 3, 4, 12, 13, 16, 17, 19–23 and 25–27. The percentage of agreement for items 1, 2, 5–11, 14, 15, 18 and 24 was between 89.5 and 94.7%. One CP-CVI experts suggested that these items could also be used for children with ocular visual impairment.

On the CVI-MQ for children with GMFCS IV and V (Table 2), the highest level of agreement (100%) among CP-CVI experts was for items 5, 7, 8, and 10–12. The percentage of agreement among experts for items 1–4, 6, 9, 13, and 14 was between 90 and 95%.

Third Delphi round

After receiving the experts' comments, we processed the proposed adaptations and resubmitted them to the two groups of experts. The usability results showed a consensus of 95% agreement on each CVI-MQ among the CP experts. The CP-CVI experts indicated 12 (5) mean (SD) minutes to administer the CVI-MQ for children with GMFCS I, II and III, and the CP experts indicated 14 (10) mean (SD) minutes to administer it (Table 1). The CP-CVI experts indicated 9 (4) mean (SD) minutes to administer the CVI-MQ for children with GMFCS IV and V, and the CP experts indicated 14 (10) mean (SD) minutes (Table 2).

Sensitivity and specificity of CVI-MQs

The MQs were completed by paediatric physical therapists and occupational therapists who were familiar with the child. We collected data from 82 children with both CP and CVI ($n = 57$ boys and $n = 25$ girls). Table 3 shows the children's characteristics. All children with CVI were included in our study and no selection was carried out based on subtypes. We therefore assumed that different subtypes are represented in our study.

The mean (SD) age in years between children with and without CVI differs 2 (4) years for GMFCS I, II and III and 1 (3–4) year for GMFCS IV and V. The number of children with CVI in the group of children with GMFCS I and V was higher compared with the group of children with GMFCS II–IV, and the number of children without CVI was in the group of children with GMFCS II, III and IV was higher compared with the group of children with GMFCS I and V (Table 3).

Table 3. Characteristics of CP children with and without CVI

Characteristic	Children with CVI	Children without CVI
Age in years, mean (SD), min-max		
GMFCS I, II and III	10 (4), 4–16	12 (4), 5–16
GMFCS IV and V	10 (3), 5–16	11 (4), 6–16
Gender, male/female (*n*, %)		
GMFCS I, II and III	11 (55)/9 (45)	16 (70)/7 (30)
GMFCS IV and V	15 (79)/4 (21)	15 (75)/5 (25)
Type of cerebral palsy (GMFCS I–V):		
spastic (*n*, %)	36 (92)	41 (95)
dyskinetic (*n*, %)	3 (8)	2 (5)
GMFCS I (*n*, %)	bilateral 11 (28), unilateral left 1 (3)	bilateral 3 (7), unilateral left 1 (2), unilateral right 4 (9)
GMFCS II (*n*, %)	2 (5) bilateral	6 (14) bilateral
GMFCS III (*n*, %)	6 (15) bilateral	9 (21) bilateral
GMFCS IV (*n*, %)	9 (23) bilateral	16 (37) bilateral
GMFCS V (*n*, %)	10 (26) bilateral	4 (9) bilateral
Speech/language development (GMFCS I–V):		
ICF-CY, d3101 = understands simple spoken messages (*n*, %)	35 (90)	42 (98)
ICF-CY, d3102 = understands complex spoken messages (*n*, %)	34 (87)	38 (88)
ICF-CY, d330 = speaks (*n*, %)	22 (56)	38 (88)
Level of intellectual disability (IQ) (GMFCS I–V): mild/moderate (*n*, %)	14 (36)/25 (64)	23 (54)/20 (46)
Presence of epilepsy (GMFCS I–V): yes/no (*n*, %)	7 (18)/32 (82)	5 (12)/38 (88)

GMFCS, gross motor function classification system; *n*, numbers; ICF-CY, International Classification of Functioning, Disability and Health, Child & Youth version (Dutch translation); IQ, intelligence quotient.

The scatter plots show the distribution of CVI-MQs scores of children with CP (Figs 1 & 2). Figure 1 shows that children without CVI and GMFCS I, II and III have a score below 10 and children with CVI and GMFCS I, II and III have a score above 10, except for one child. Figure 2 shows that children without CVI and GMFCS IV and V have a score below 8 and children with CVI and GMFCS IV and V have a score above 8. A cut-off score of 12 or higher (Fig. 1, Table 4) indicates probability of presence of CVI in a child with CP and GMFCS I-II-III. A cut-off score of 8 or higher indicates probability of presence of CVI in a child with CP and GMFCS IV-V (Fig. 2, Table 4). Table 4 presents the values of sensitivity and specificity and corresponding cut-off scores for both CVI-MQs.

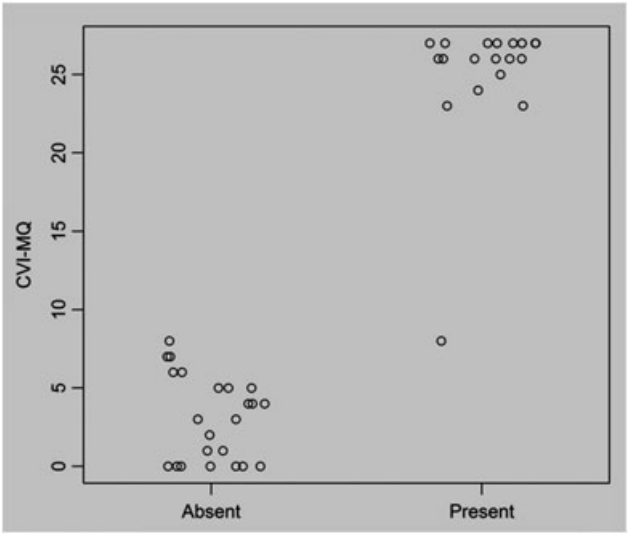

Figure 1. Scatter plot of CVI-MQ scores for children with GMFCS I, II and III. Absent, CVI is absent; Present, CVI is present; CVI-MQ, Cerebral Visual Impairment Motor Questionnaire for children with CP; GMFCS, gross motor function classification system.

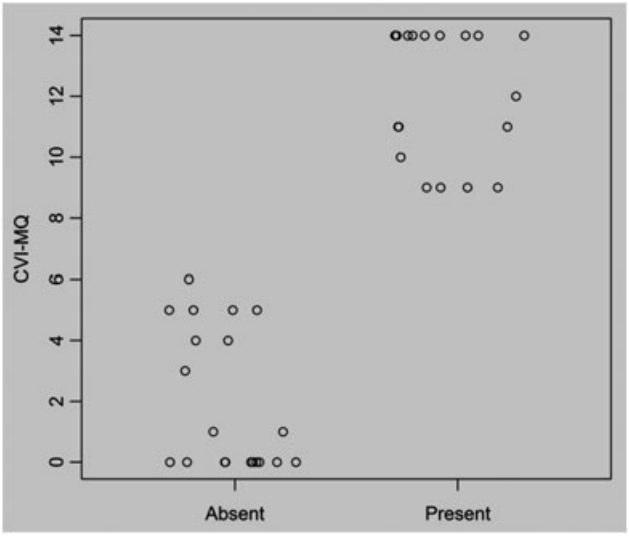

Figure 2. Scatter plot of CVI-MQ scores for children with GMFCS IV and V. Absent, CVI is absent; Present, CVI is present; CVI-MQ, Cerebral Visual Impairment Motor Questionnaire for children with CP; GMFCS, gross motor function classification system.

Tables 1 and 2 show the frequency in percentage answers item (Yes, No and Not applicable) for each item on the both CVI-MQs, which the experts filled in on the motor behaviour of the children. A 'Yes' points to the probability of CVI and a 'No' does not. In the scoring system every 'Yes' means 1 point and every 'No' means 0 point on the scale. An item was evaluated 'not applicable' by the professional if a child was considered of being too old or too young in order to perform a certain motor skill, or that the motor skill was too difficult to perform in relation to the child's GMFCS level. Such is interpreted as a score of zero as presence is not observed. This procedure makes it possible to assign a total score for each child in Tables 1 and 2 as the number of present motor skills.

The results of the ROC curve for CVI-MQ, GMFCS levels I, II and III are sensitivity 1.00 and specificity 0.96, and for CVI-MQ, GMFCS levels IV and V sensitivity 0.97 and specificity 0.98. This indicates excellent sensitivity and specificity for identifying at-risk children with CP with the possibility of having CVI.

Figure 1 shows that among the children with CVI, one child (GMFCS I) received a lower sum score on the CVI-MQ for children with GMFCS I, II and III, whereas all other children clearly received a higher CVI-MQ score. This child has GMFCS level I and mild level of intellectual disability.

Taking 7.5 as the cut-off score for GMFCS I, II and III corresponds to a sensitivity rate of 1.00 and a specificity rate of at 0.96, or 15.5 points and higher if the sensitivity rate is 0.95 and the specificity rate 1.00. We therefore chose a score of 12 as cut-off value in order to meet a maximal sensitivity and specificity for children with GMFCS levels I, II and III.

Because the sum scores of the groups with and without CVI are completely separated in Fig. 2, a ROC curve estimation procedure would indicate an area under the curve equal to one. For the ROC in Fig. 1, this differs because there was a single child observed with a lower CVI-MQ score. The corresponding area under the curve obtained was 0.99 with a 95% confidence interval equal to 0.99–1.00.

Discussion

The two CVI-MQs for children with CP have good face validity and are usable tools to detect children suspected of having CVI. These questionnaires have excellent sensitivity and specificity as well as a positive/negative predictive value with feasible cut-off scores.

During the Delphi rounds, the CP-CVI experts suggested several issues that may influence validity. First, the difficulty with moving in an unfamiliar environment compared with a familiar environment (items 4, 6–8, 11, 14, 15, Table 1; item 6, Table 2) could be due to not feeling safe/secure enough. On the other hand, moving without difficulty in a familiar environment could be the result of automated motor patterns rather than the familiarity of the environment (Cohen-Maitre and Haerich, 2005).

The questionnaire also includes some complex tasks (item 20, Table 1; and item 7, Table 2), caused, for example, by difficulty with depth perception, distance viewing or absence of visually guided reach (item 20, Table 1). With respect to item

Table 4. Sensitivity and specificity values and cut-off scores for the two CVI-MQs

	GMFCS I, II and III	GMFCS IV and V
Cut-off scores	12	8
Sensitivity (point estimates and 95% CI)	1.00 (0.76–1.00)	0.97 (0.79–1.00)
Specificity (point estimates and 95% CI)	0.96 (0.78–1.00)	0.98 (0.80–1.00)
Area under the curve (AUC) value	0.99	1.00
Standard error	0.002	0.000
Positive predictive value (point estimates and 95% CI)	0.95 (0.76–1.00)	0.97 (0.79–1.00)
Negative predictive value (point estimates and 95% CI)	1.00 (0.78–1.00)	0.98 (0.80–1.00)
Positive likelihood ratio (point estimates and 95% CI)	23.00 (3.38–156.39)	40.95 (2.65–633.88)
Negative likelihood ratio (point estimates and 95% CI)	0.00 (0.00–0.00)	0.03 (0.00–0.40)
Asymptomatic significance	<0.001	<0.001
Asymptomatic 95% CI (lower bound–upper bound)	0.000–0.006	0.000–0.000

CI, confidence interval; GMFCS, gross motor function classification system.

19 (Table 1), the difficulty is caused not only by the child moving but also by changes in the environment.

The motor development of children with visual impairment is qualitatively and quantitatively different compared with children with normal vision. This is true for both the fine-motor skills as well as for their gross motor abilities. Recent reviews in children with developmental coordination disorder (DCD) (Adams *et al.* 2014) and CP (Steenbergen *et al.* 2013) have suggested that the motor deficits observed in these children have a common origin in a deficit in internal models of motor control. Clearly, visual-motor function in children with DCD and CP depends on intact visual and motor systems and their interaction. The extent to which this interaction affects motor functioning in CP warrants further study. For items 21 and 22 (Table 1), it is important to assess if the child uses visual guidance before reaching for a small object, which could be a result of visual support. When the child adapts the size of its hand to the size of the object after touching it, it could be a result of tactile support rather than obtained visual information. In the item 'The child does not reach for and look at an object at the same time' (item 26, Table 1), the presence of CVI could affect serial processing in the brain, resulting in difficulty with multitasking (looking and reaching at the same time).

In Table 1 (item 1, 2) and Table 2 (item 1, 2, 3, 4 and 13), we used the word 'if' in order to emphasize that without sound production, fluorescence*, high-contrast* toys or verbal stimulation, the child will have difficulty to perform the motor task.

In our study, children aged of 4 to 16 years are included. The CVI-MQ for children with GMFCS I, II and III shows that some questions (e.g. 4, 13, 19) may be age-dependent. For instance, a 4-year-old child without CVI may not be able to estimate distance and speed of other road users because of his/her young age. Figures 3 and 4 show the range of ages in children. Because most of children are older than 6 years, we assume that the age of children does not affect the results of our study.

Early detection of developmental problems such as CVI is needed for a professional to facilitate an early start in appropriate intervention for these children and support for their parents. This has been proven to be beneficial and improves outcome (Malkowicz *et al.* 2006). The professional who is familiar with the child with CP filled in the questionnaire based on his or her knowledge on that child. Therefore, it is not necessary that the child was present during this procedure. Using the CVI-MQs enables to quickly achieve information on the risk of CVI in children with CP. When a child with CP is able to perform a motor skill during the therapy but the results of a motor test do not show it or when a child needs extra verbal and manual support to accomplish a motor task, it is recommended to use the CVI-MQ. Using these screening tools can also help paediatric physical therapists and occupational therapists to assess children with CP when additional certainty is desired about whether the current impairments of a child with CP are not only caused by motor or mental delay but perhaps also by the presence of CVI.

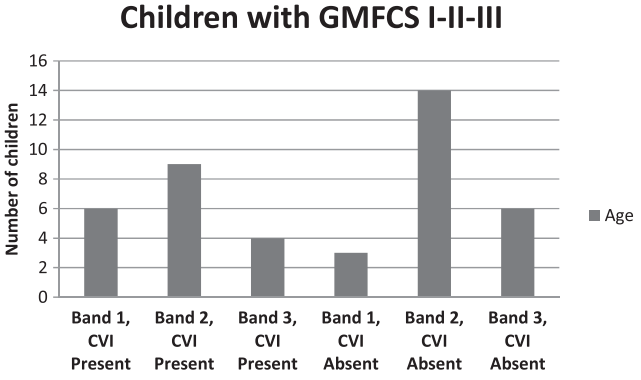

Figure 3. Age of children with gross motor function classification (GMFCS) level I, II and III. X-axis: children with (Present) and without (Absent) CVI in three different range of age in years, Y-axis: number of children. Band 1, children ages 4–6; band 2, children ages 6–12; band 3, children ages 12–16.

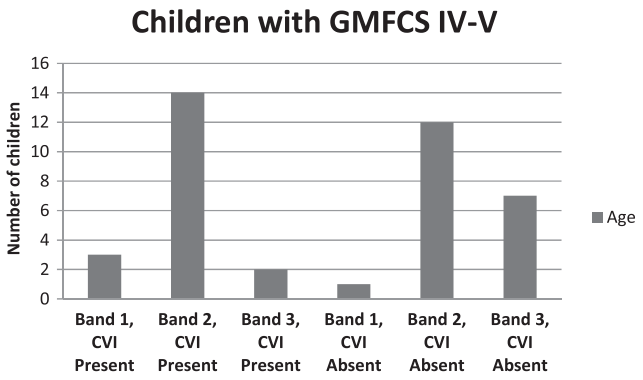

Figure 4. Age of children with gross motor function classification (GMFCS) level IV and V. X-axis: children with (Present) and without (Absent) CVI in three different range of age in years, Y-axis: number of children. Band 1, children ages 4–6, band 2; children ages 6–12; band 3, children ages 12–16.

Presuming the presence of CVI as a result of a positive score on the CVI-MQs could be the first step towards a timely and accurate diagnostic for a child with CP. In the absence of red flags, it also prevents unnecessary comprehensive testing of children and is cost-efficient and time-efficient. Use of these CVI-MQs for children with CP is therefore relevant and warranted.

The professionals familiar with motor screening can use CVI-MQ to screen children with CP in approximately 10 min to predict presence of CVI in children and to refer for further assessment. We recommend using the CVI-MQs as a part of comprehensive research in addition to other screening tools for CVI.

It should be noted that the content of both CVI-MQs consists almost entirely of items at the level of motor functioning related to depth perception. CVI could result in, for example, a strong colour preference, visual latency-delayed responses in looking at objects, difficulties with visual complexity, light-gazing and non-purposeful gaze, absent or atypical visual reflexes (Dutton and Jacobson, 2001; Stiers *et al.* 2002). To investigate these characteristics of CVI it is important to use the available CVI screening tools (Dutton and Jacobson, 2001; Steendam, 2008; Ortibus *et al.* 2011; Dutton *et al.* 2012).

Limitations

In the current study, the experts had prior knowledge of the presence of absence of CVI in children with CP; this may have affected the results of sensitivity and specificity. We recommend blind scoring in future studies.

We received the CVI diagnose from healthcare centres without additional information about visual characteristics of the participants. This is a limitation in the interpretability of the results. In the future studies, it is important to obtain a more full report of the characteristics of CVI.

Conclusion

The CVI-MQs are a valuable addition for paediatric physical therapists and occupational therapists working with children with CP to detect the presence of CVI. Implementing CVI-MQs as part of clinical reasoning is important in order to screen children with CP and identify red flags for CVI.

Key messages

- CVI can result in a delayed motor development in children with CP.
- The red flags allow professionals to review the impact of CVI on the observed motor behaviour in children with CP.
- The CVI-MQs are a valuable addition for professionals working with children with CP to detect presence of CVI.

Conflict of interest

The authors declare that there is no conflict of interest.

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